the epidural space for 11 days, which is the longest recorded period for this technique. The reported principal hazards of continuous epidural catheter techniques have been epidural infection[4] and catheter breakage.[5] Neither occurred in this case.

## Summary

A case of acute herpes zoster was successfully treated by continuous epidural analgesia. The catheter was left in place for 11 days without infection.

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# Occult Neoplasm Causing Syndrome of Retroperitoneal Fibrosis

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RETROPERITONEAL FIBROSIS is one name for a syndrome which includes the gradual obstruction of one or both ureters by a hard fibrous plaque which is often, although not always, limited to the periureteral regions. The process has also been called *periureteritis plastica, periureteritis obliterans, perirenal fascitis, periureteral lymphangitis, periureteral liposclerosis, Gerota's fascitis, retroperitoneal adiponecrosis, idiopathic non-specific fibrosing retroperitonitis* and *Ormond's syndrome.* As is usually the case when a disease is endowed with a myriad of descriptive tags, the pathologic history is obscure. Many efforts have been made to find a cause for this disease, but the more than 125 cases thus far reported resist classification by pathogenesis.

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Certain neoplastic diseases which are sometimes associated with pronounced fibrotic reaction have been suggested as causative factors. These include Hodgkin's disease and other lymphomas, certain sarcomas and anaplastic carcinomas that spread to the retroperitoneal space. Post traumatic or post surgical scarring has been impugned as an etiologic possibility. When the retroperitoneal inflammation includes fatty tissue and takes a form of a granulomatous reaction, the process has been called *sclerosing lipogranuloma.* Retroperitoneal fibrosis has been considered as part of a generalized hypersensitivity phenomenon as seen in periarteritis nodosa or Wegener's arteritis. Severe urinary tract infection of long standing has been suggested as a cause, as have actinomycosis and other fungus infections. In some cases there has been an associated inflammatory condition of the bowel, pelvis or lower extremity. Some patients have had aortitis. In others, lymphangitis (or at least involvement of the lymphatics with edema of the lower extremity) has been a prominent feature. Experimentally, the presence of urine in the retroperitoneal space has been shown to cause fibrotic reaction. Syphilis classically produces fibrotic tissue but in only one of the cases reviewed was positive reaction to a serologic test for syphilis reported. Recent reports indicate that the anti-

serotonin agent methysergide maleate (Sansert®), is the one known etiologic agent.

Unfortunately, pathologic examination of the fibrous plaque has not been too helpful. The cellular component is variable and often scant. Generalized vasculitis is usually seen but it is non-specific. The prominent feature is firm uniform fibroblastic proliferation which is found behind the peritoneum on either or both sides as far lateral as the lumbar gutters. It has been found as low as the level of the trigone and as high as the diaphragmatic crura.

## Reports of Cases

CASE 1. A 29-year-old white machinist was first seen on December 12, 1963 with acute left ureteral colic. Except for a low backache of two weeks' duration he said he had no pains. His general health had been excellent. He had not had fever, chills, nausea or frequency of urination. No masses were palpable in the abdomen and there was no tenderness in either costovertebral angle or in the abdomen. No muscle guarding was evident but a left psoas sign was equivocally positive. No lymph nodes were palpable and no hernia was present. External genitalia were normal. The rectal sphincter was thickened as the result of a previous operation for a perirectal abscess. The prostate gland was normal in size and texture and no abnormal rectal masses were felt. Results of urinalysis were within normal limits. An excretory urogram showed promptly functioning kidneys of equal size and without dilatation. In retrospect the ureters were slightly deviated medially on these films. Tentative diagnosis was made of a passed left ureteral calculus.

Eight weeks later the patient was seen again, complaining of progressively severe pain in the left lower abdominal quadrant, radiating down the backs of both thighs. He required increasing amounts of morphine. He was weak and pale without signs of overt bleeding. He said he had had no difficulty in evacuation of bowel or bladder. Physical examination was unchanged except for pallor and mild spasm of the paravertebral muscles. Results of urinalysis again were within normal limits. No abnormalities were seen in roentgenograms of his chest. Hemoglobin, which had been 14.5 gm per 100 ml at the time of the previous examination, now was 9.0 gm. Erythrocyte sedimentation rate was 115 mm in one hour (Wintrobe). An excretory urogram demonstrated no function on the right in two hours and minimal left caliectasis without pyelectasis. The bladder outline appeared normal and the bladder emptied well, judging from the post voiding film.

Cystoscopic examination and retrograde pyelograms were carried out on February 11, 1964. The bladder and urethra were normal. A ureteral catheter passed easily to the left kidney and pyelograms confirmed the presence of mild caliectasis. The right ureteral catheter met mild resistance approximately 6 cm from the bladder, but then passed easily to the kidney, which was evacuated of 45 ml of clear urine. Right retrograde pyelograms revealed moderate hydronephrosis. The right ureteral catheter was left in place and splinted with an indwelling urethral catheter. In the differential diagnosis retroperitoneal fibrosis and a retroperitoneal malignant lesion involving both ureters were considered.

Transperitoneal exploration was performed February 13, 1964 through a midline incision. A large plaque of hard smooth fibrous material encased the ureter on the right and extended from below the sacral promontory to the renal arteries. To the left of the midline a narrow tongue of fibrous tissue extended across the aorta, partially encasing the left ureter. The right ureter was freed from the mass with difficulty while the left was easily freed from its relatively superficial involvement. Both ureters were transplanted intraperitoneally. A generous biopsy specimen was taken from the mass near the level of the fifth lumbar vertebra on the right, permitting some necrotic material to ooze from the center of the plaque. Microscopic examination revealed "non-specific inflammation with fibrosis." The slides were examined by two oncologists who agreed regarding the benignity of the lesion. Routine cultures from the caseous material produced no growth.

The patient's immediate postoperative course was ordinary. He regained strength and pain was not prominent. He was given a total dose of 900r of x-irradiation to both lumbar gutters from midsacrum to diaphragm.

A month later he began to have severe pain in both flanks which radiated anteriorly around the pelvic girdle and into the subcostal regions. Roentgenograms of the chest showed decided widening of the upper mediastinum. X-irradiation was resumed and extended to include the mediastinum. In addition the patient was given dexamethasone 0.5 mg 3 times daily. Dramatic relief followed and the patient returned to work in 48 hours. However,

debilitating flank and pelvic pain soon recurred.

The patient was readmitted to the hospital where roentgenograms of the chest showed decided extension of the mediastinal mass into the upper lung fields. Biopsy of enlarged lymph nodes felt in the left supracervical region showed reticulum cell sarcoma. The patient died June 24, 1964 with widely disseminated tumor.

CASE 2. A 57-year-old woman of Mexican extraction was examined by an internist in September 1962 for weakness and anemia of unknown cause. She complained that eating large meals produced excessive epigastric fullness. She had had no loss of weight, no urinary difficulty, no abdominal pain and no hematuria. Previous operations included a hysterectomy for benign fibromyomata 10 years previously and appendectomy five years before that. On physical examination the only abnormality noted was a smooth, fixed, non-tender mass approximately 6 cm × 12 cm deep in the left mid-abdomen. It was not palpable through the vagina or rectum. Hemoglobin was 11.9 gm per 100 ml of blood. Leukocytes numbered 10,250 per cu mm and the cell differential was within normal limits. Erythrocyte sedimentation rate was 29 mm in one hour (Wintrobe). An excretory urogram revealed non-function of the left kidney. No abnormalities were seen in roentgenograms of the intestinal tract and chest. Cystoscopic examination and left retrograde pyelograms were carried out September 19, 1962. The bladder and urethra appeared normal. A ureteral catheter was passed to the left kidney with ease but a left ureterogram revealed pronounced narrowing of the left mid-ureter for approximately 8 cm. Urine specimens from the left kidney and bladder were negative for malignant cells. No growth resulted from routine cultures. The left kidney showed slight atrophy and no hydronephrosis. The presumptive diagnosis was retroperitoneal fibrosis limited to the left side.

On September 24, 1962 a left lumbar incision was made and a thick, white, fibrous mass was seen forming a sheath about the left ureter from the renal pedicle to the level of the bifurcation of the common iliac artery. The kidney appeared normal. The peritoneum was easily separated from the mass and the left ureter was transposed to an extraperitoneal position. Biopsy of the fibrous mass revealed "non-specific inflammation with fibrosis." Postoperative recovery was normal, but gradually pain developed in the left flank. Six weeks after operation an excretory urogram showed no return of left renal function in four hours. On retrograde studies the left ureter was seen to be patient. Radioactive renograms demonstrated no function on that side. Because of progressive left abdominal pain, x-irradiation amounting to 1150 r in air was given during a two-week period. The left abdominal mass diminished and became non-palpable. Pain persisted and the left kidney did not return to function. On April 1, 1963 left nephrectomy was carried out. At operation the fibrotic process was seen to involve the renal pedicle but it showed no sign of extending across the midline. No abnormalities were noted on peritoneal exploration. The removed kidney showed chronic non-specific phelonephritis with atrophy. The patient recovered and pain was relieved.

On the 14th postoperative day, the surgeon noted a 2×3 cm mass on the left side of the patient's neck. It was soft and movable and felt like an enlarged lymph node. She was examined by a general surgeon and excisional biopsy was scheduled two weeks later but by that time the mass had disappeared and the biopsy was canceled.

Approximately a year and a half after ureterolysis and a year after nephrectomy the patient brought to our attention a small hernia through the lumbar scar. An excretory urogram showed a hypertrophied but otherwise normal right kidney. The patient had no pain but wished to have the hernia repaired.

As it happened, it was at this time that the young machinist in Case 1 was found to have malignant disease and we recalled the peculiarly transient node in this woman's neck. On careful reexamination no lymph nodes were felt in her neck or elsewhere. Nonetheless, at the time of lumbar herniorrhaphy on April 16, 1964 nodes were removed from the left scalene fat pad and on biopsy giant cell lymphosarcoma was diagnosed.

## Discussion

The delayed finding of malignant disease in these two cases led us to explore the literature and carry out personal communication in an attempt to ascertain the frequency of this situation. We were impressed by the lack of documented follow-up in cases of so-called "benign idiopathic retroperitoneal fibrosis." We would like to know if five-year follow-up will alter the 7 to 16 per cent mortality rates now reported in the literature.

As discussed above, the cause and exact nature of this disease are poorly understood and highly

speculative. It is commonly considered a benign entity and discussed as such.

In the two patients reported upon herein, careful gross and microscopic examinations were made, the disease was deemed benign and, therefore, presumably the patients were cured or at least assured of a reasonable prognosis by adequate surgical treatment. One is dead from a reticulum cell sarcoma which in retrospect was in part (or entirely) the original process. The other patient is now known to have a sarcoma but ultimate evaluation of its relationship to the original retroperitoneal findings must await autopsy.

The fortuitous finding a lymphoma in the second case by scalene biopsy prompts us to recommend that such biopsy be a part of the evaluation of all patients with suspected retroperitoneal fibrosis.

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# An Unusual "Thyroid" Nodule "Cold" to Scintiscan

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RADIOISOTOPE SCINTISCANNING is widely employed in the clinical evaluation of thyroid nodules. When a nonfunctioning ("cold") solitary nodule is demonstrated, potential thyroid malignant disease is suspected. The incidence of confirmed carcinoma in such cold nodules varies and has been reported to be as high as 55 per cent.[2]

Various benign lesions will result in the appearance of a cold nodule on scintiscanning. These include cystic changes, degeneration, necrosis, hemorrhage and nonfunctioning adenomas. In the following case report, another type of benign lesion is described, which produced a solitary cold area on thyroid scintiscan.

## Report of a Case

A 58-year-old Caucasian woman was admitted to the University of California San Francisco Medical Center on August 2, 1964. Except for intermittent joint discomfort attributed to arthritis, she had been in apparent good health until June 1964. At that time she had noted sudden onset of severe pain in the region of the right groin. Two weeks later roentgenograms of the right femur revealed an osteolytic lesion in the neck and another in the shaft. A metastatic carcinoma was suspected and the patient was admitted to another hospital for further study on July 13, 1964.

On the initial admission, mild hypertension was noted (blood pressure 170/90 mm of mercury). The thyroid gland was diffusely enlarged. The right lobe was larger than the left and it contained a 2 x 2 cm mass in the midportion. Moderate kyphoscoliosis was observed and pain was elicited on movement of right hip. The serum calcium level was

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